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Role of myringotomy in patient with cleft palate: Meta-analysis study.

Ahmed Taha Sayed Hassan

Professor of Plastic surgery & burn and reconstructive surgery department,
Faculty of Medicine, Al-Azhar University (Cairo), Egypt

Ahmed Abu El-Soud Salim

Assistant professor Plastic surgery & burn and reconstructive surgery
department, Faculty of Medicine, Al-Azhar University (Assiut), Egypt

Asmaa Ali Dahy

Lecture of Plastic surgery & burn and reconstructive surgery department, Faculty of
Medicine, Al-Azhar University (Cairo, girls), Egypt

Mahmoud Abdelhamid Abdelwahab Elhendawy

Lecture of Plastic surgery & burn and reconstructive surgery department, Faculty of
Medicine, Al-Azhar University (Assiut), Egypt

Mohamad Ismail Elkady Mohamed

Resident of plastic surgery, Egypt

Corresponding author email: drmohamedelkady665@gmail.com

Abstract---Background: Cleft lip and palate (CLP) are among the most common congenital malformations. Patients with CLP have higher rates of health complications such as speech delays, behavioral disorders, failure to thrive, otitis media, hearing loss. Aim and objectives: the aim of the study was to evaluate speech and audiologic outcomes after palatoplasty with or without myringotomy (with or without ventilation tube) in children with cleft palate through a meta- analysis study from 10 years ago. Subjects and methods: A literature search was performed in PubMed, PLOS and Clarivate - Scopus listed articles. We will use many research engines including EKB to download the articles for the last one year. Results: Our results showed that 3 studies showed fistula Event rate (%) 8.482% with significant heterogeneity p-value 0.0230. 2 studies showed OM Event rate (%) 17.35% with significant heterogeneity p-value 0.0084. The current study showed that 2 studies showed poor hearing with tube Event rate (%) 43.327% with insignificant heterogeneity p-value 0.1278. Conclusion: We have produced a recommendation about the outcomes that should be measured, as a minimum, in studies of the management of OME in children with CP. The development process included input from key stakeholders and used novel methodology to integrate the opinion of healthcare professionals, parents and children.

Keywords---cleft lip, palate, otitis media, speech delays, myringotomy, tympanostomy.

Introduction

Cleft lip and palate (CLP) are among the most common congenital malformations. Patients with CLP have higher rates of health complications such as speech delays, behavioral disorders, failure to thrive, otitis media, hearing loss, and dental anomalies, for which they undergo numerous surgical procedures.¹ Although operative treatment has been proven to be beneficial for these patients, there are increasing concerns about higher exposure to general anesthesia and its association with abnormal neurocognitive development.²

A common comorbidity seen in children with CLP is recurrent otitis media, with a reported prevalence of 97%, primarily due to concomitant Eustachian tube dysfunction, when left untreated, chronic ear dysfunction often causes atrophy and scarring of the tympanic membrane, resulting in conductive hearing loss.³ The association between cleft palate and otitis media with (OM), hearing loss and chronic otitis media (COM) is well documented.⁴ Otitis media (OM) is reported to be almost universal among infants with cleft palate.⁵

Ventilation tube insertion is a safe and effective way to relief middle ear dysfunction and audition loss. It should be considered to be a basic principle of therapy.⁶ Myringotomy and insertion of a tympanostomy tube are performed to release middle ear fluid in pediatric patients with chronic otitis media (COM). Drainage of effusion in the middle ear via a small incision in the tympanic membrane leads to resolution of COM.⁷ The study aimed to evaluate speech and audiologic outcomes after palatoplasty with or without myringotomy (with or without ventilation tube) in children with cleft palate though a meta-analysis study from 10 years ago.

Patients and Methods

A literature search was performed in PubMed, PLOS and Clarivate -Scopus listedarticles. We will use many research engines including EKB to download the articles for the last one year. All published studies investigating the role of myringotomy in patient with Cleft Palate.

Two independent reviewers will do data extraction systematically, adhering to the PRISMA guidelines. Summary measures will pool in a random-effects model meta-analysis. Study was assessed type of cleft, types of tube insertion, number of tube, speech and hearing outcome, the incidence of tube related complications in this meta-analysis.

Results

Author	type of study
Park MS et al., ⁸	retrospective
Sumun K et al., ⁹	retrospective
Jung SE et al., ¹⁰	retrospective
Mantilla-Rivas et al., ¹¹	retrospective
Shaffer AD et al., ¹²	retrospective
Yang CH et al., ¹³	retrospective

Bruneel L et al., ¹⁴	retrospective
Imbery TE et al., ¹⁵	retrospective
Kim E et al., ¹⁶	retrospective
Ha S et al., ¹⁷	retrospective
Annigeri VM et al., ¹⁸	retropective
Kobayashi H et al., ⁵	retopective

Table 1: Study characteristics

12 studies were included all were retrospective studies as shown in table 1

author	number	age	m\ f
Park MS et al., ⁸	636		386\250
Sumun K et al., ⁹	19		6\13
Jung SE et al., ¹⁰	72	2.8	38\34
Mantilla-Rivas et al., ¹¹	74	10.6	38\36
Shaffer AD et al., ¹²	232		149\83
Yang CH et al., ¹³	90	1.15	55\35
Bruneel L et al., ¹⁴	16	5.4	6\10
Imbery TE et al., ¹⁵	352	1	
Kim E et al., ¹⁶	147		96\51
Ha S et al., ¹⁷	292	6.04	147\145
Annigeri VM et al., ¹⁸	30	2.7	15\15
Kobayashi H et al., ⁵	133	9.42	72\61

Table2. Patient's characteristics

A total cases included were 2093 m\ f was 1008\ 733 and mean age was 4.8 years as shown in table2

Study	Total number	Event	Event rate (%) (Proportion)	95% CI of rate (%)
Jung SE et al., ¹⁰	72	19	26.389	16.695 - 38.103
Shaffer AD et al., ¹²	232	65	28.017	22.338 - 34.268
Bruneel L et al., ¹⁴	16	13	81.25	54.354 - 95.953
Total (fixed effects)	320		30.383	25.413 - 35.717
Total (random effects)	320		41.304	21.379 - 62.846
Test for heterogeneity				
Q	19.2399			
DF	2			
Significance level	0.0001*			
I ² (inconsistency)	89.60%			
95% CI for I ²	71.96 - 96.15			

Q: Total variance for heterogeneity

I²: Observed variance for heterogeneity

CI: Confidence interval (LL: Lower limit-UL: Upper Limit)

Table (3): Meta-analysis for Nasal emission

3 studies showed Nasal emission Event rate (%) (Proportion) 41.304% with significant heterogeneity p-value 0.0001

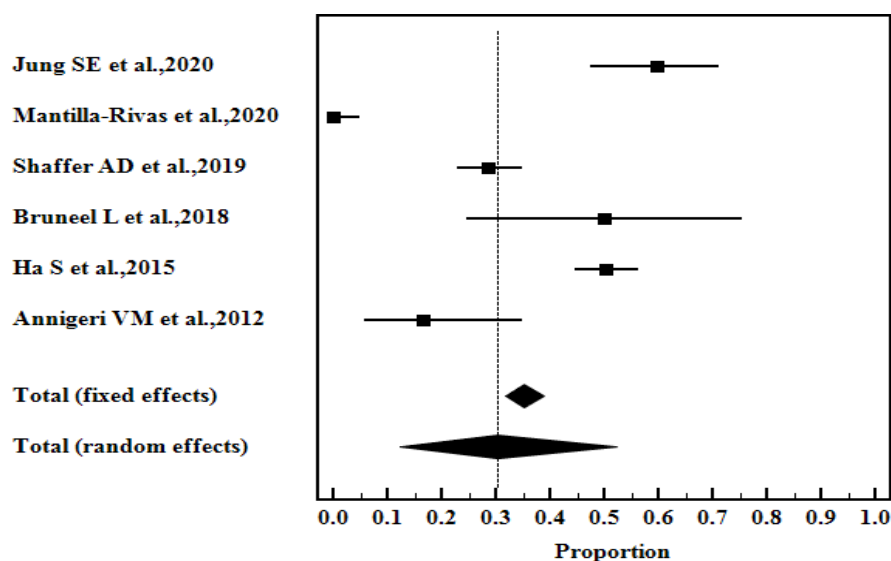


Figure (1): Forest plot for speech therapy

6 studies showed speech therapy Event rate (%) (Proportion) 30.348% with significant heterogeneity p-value 0.0001

Study	Total number	Event	Event rate (%)	95% CI of rate (%)
Jung SE et al., ¹⁰	72	28	38.889	27.615 - 51.106
Shaffer AD et al., ¹²	232	70	30.172	24.338 - 36.521
Ha S et al., ¹⁷	292	84	28.767	23.643 - 34.328
Total (fixed effects)	596	84	30.595	26.924 - 34.458
Total (random effects)	596	84	30.954	26.529 - 35.561
Test for heterogeneity				
Q	2.7469			
DF	2			
Significance level	0.2532			
I ² (inconsistency)	27.19%			
95% CI for I ²	0.00 - 97.56			

Q: Total variance for heterogeneity

I²: Observed variance for heterogeneity

CI: Confidence interval (LL: Lower limit-UL: Upper Limit)

Perforation

Table (4): Meta-analysis for hyper-nasality

3 studies showed hyper-nasality Event rate (%) (Proportion) 30.954% with insignificant heterogeneity p-value 0.2532

Study	Total number	Event	Event rate (%)	95% CI of rate (%)
Imbery TE et al., ¹⁵	352	87	24.716	20.297 - 29.566
Kim E et al., ¹⁶	147	50	34.014	26.410 - 42.276

Kobayashi H et al., ⁵	133	5	3.759	1.232 - 8.555
Total (fixed effects)	632		21.169	18.053 - 24.556
Total (random effects)	632		18.967	5.329 - 38.393
Test for heterogeneity				
Q	55.5134			
DF	2			
Significance level	<0.0001*			
I ² (inconsistency)	96.40%			
95% CI for I ²	92.45 - 98.28			

Q: Total variance for heterogeneity

I²: Observed variance for heterogeneity

CI: Confidence interval (LL: Lower limit–UL: Upper Limit)

Table (5): Meta-analysis for Perforation

3 studies showed Perforation Event rate (%) (Proportion) 18.967% with significant heterogeneity p-value <0.0001

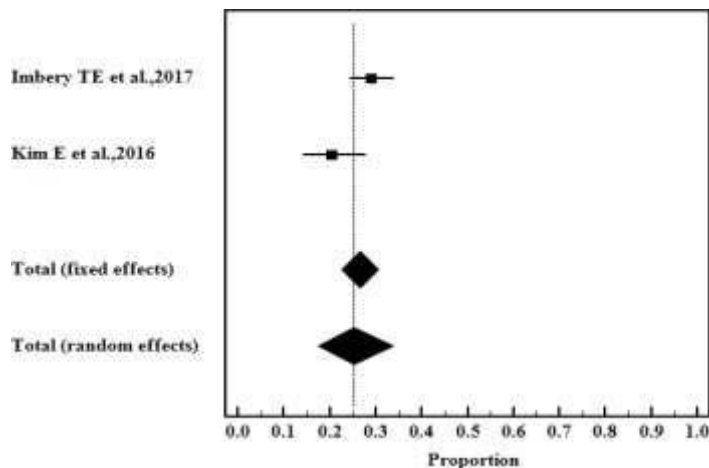


Figure (2): Forest plot for Myringosclerosis

3 studies showed Myringosclerosis Event rate (%) (Proportion) 25.134% with significant heterogeneity p-value 0.0456

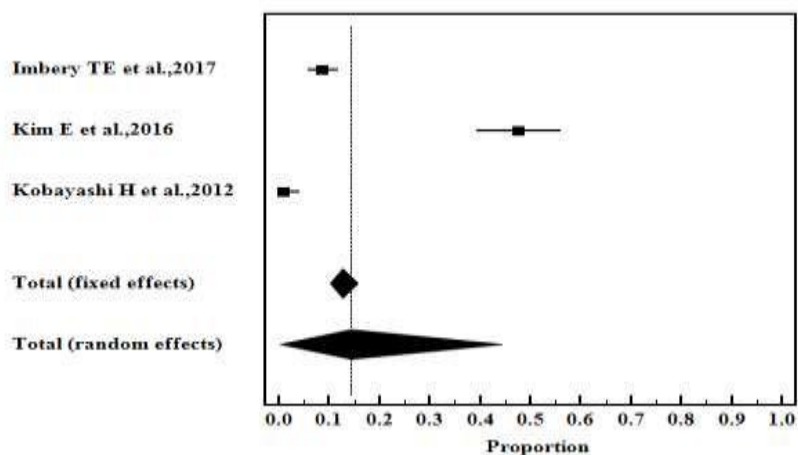


Figure (3): Forest plot for Retraction

3 studies showed Retraction Event rate (%) (Proportion) 14.317% with significant heterogeneity p -value <0.0001

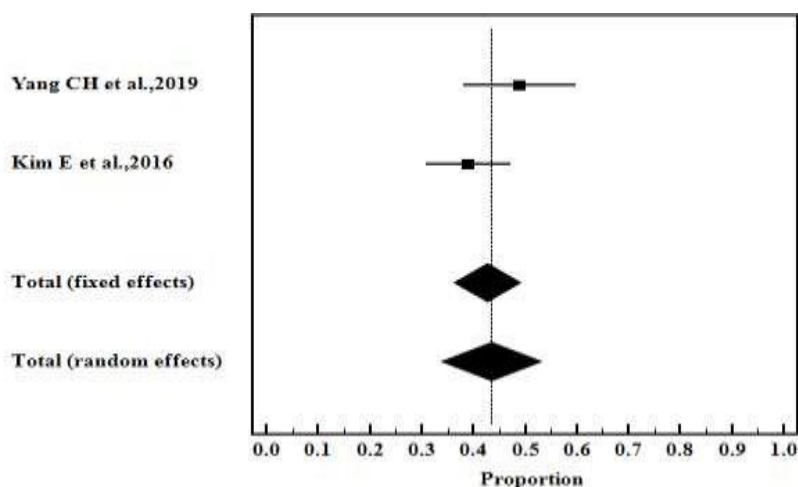


Figure (4): Forest plot for poor hearing with tube

2 studies showed poor hearing with tube Event rate (%) (Proportion) 43.327% with insignificant heterogeneity p -value 0.1278

Study	Total number	Event	Event rate (%) (Proportion)	95% CI of rate (%)
Jung SE et al., ¹⁰	72	39	54.167	42.004 - 65.975
Ha S et al., ¹⁷	292	150	51.37	45.477 - 57.234
Kobayashi H et al., ⁵	133	27	20.301	13.825 - 28.145
Total (fixed effects)	497		42.975	38.589 - 47.445
Total (random effects)	497		41.288	20.785 - 63.516
Test for heterogeneity				

Q	44.0521
DF	2
Significance level	<0.0001*
I ² (inconsistency)	95.46%
95% CI for I ²	89.98 to 97.94

Q: Total variance for heterogeneity

I²: Observed variance for heterogeneity

CI: Confidence interval (LL: Lower limit–UL: Upper Limit)

Table (6): Meta-analysis for Articulatory deficits

3 studies showed Articulatory deficits Event rate (%) (Proportion) 41.288% with significant heterogeneity p-value <0.0001

Study	Total number	Event	Event rate (%) (Proportion)	95% CI of rate (%)
Annigeri VM et al., ¹⁸	30	2	6.667	0.818 - 22.074
Kobayashi H et al., ⁵	133	36	27.068	19.733 - 35.453
Total (fixed effects)	163		22.965	16.784 - 30.142
Total (random effects)	163		17.35	2.978 - 40.110

Test for heterogeneity	
Q	6.9392
DF	1
Significance level	0.0084*
I ² (inconsistency)	85.59%
95% CI for I ²	41.96 - 96.42

Q: Total variance for heterogeneity

I²: Observed variance for heterogeneity

CI: Confidence interval (LL: Lower limit–UL: Upper Limit)

Table (7): Meta-analysis for OM

2 studies showed OM Event rate (%) (Proportion) 17.35% with significant heterogeneity p-value 0.0084

Discussion

This metanalysis study included 10 studies were included all were retrospective studies. A total case included were 1438, m\|f was 616\ 470 and mean age was 4.8. Our results were supported by study of Yang et al.,¹³ as they reported that a total of 90 children with cleft lip and palate (55 boys and 35 girls) who underwent palatoplasty and ventilation tube insertion were enrolled in this study. The average age of children with cleft lip and palate undergoing pure tone audiometry was 6.4 years.

In the study of Ha et al.,¹⁷ they included 292 patients with non-syndromic overt cleft palate ($\pm$ cleft lip) who received primary palatal surgery. The patients included 147 boys and 145 girls and ranged in age from 36 months to 19 years 4 months (mean = 72.48 months) at the time of data collection. The present study showed

that as regard side was mainly either bilateral cleft lip and palate (250), unilateral cleft lip and palate (284), isolated cleft palate (74), a cleft palate (CP) only (161), type of cleft was Submucous cleft palate, non-Syndromic, Syndromic.

Our results were supported by study of Mantilla-Rivas et al.,¹¹ as they reported that a portion of patients (56.1%) had isolated cleft palate (complete = 15; incomplete = 35), 39 (43.8%) patients had CLP for which 13 (33.3%) were bilateral and 26 (66.7%) unilateral. Fifteen patients had a syndromic diagnosis including Stickler syndrome (n = 4), Van der Woude syndrome (n = 2), 22q11.2 deletion syndrome (n = 1), Culler-Jones syndrome (n = 1), SATB2-associated syndrome (n = 1), Kabuki syndrome (n = 1), caudal regression syndrome (n = 1), 1q22 micro duplication (n = 1), and unknown cause or other chromosomal abnormality (n = 3).

In the study of Shaffer et al.,¹² seventy-four (31.9%) children had isolated CP, 107 (46.1%) had unilateral cleft lip and palate, and 51 (22.0%) had bilateral cleft lip and palate. Also, Bruneel et al.,¹⁴ demonstrated that eight patients had a unilateral cleft lip and palate (UCLP), 4 patients presented with a bilateral cleft lip and palate (BCLP) and four patients with a cleft palate (CP) only. In the study in our hands, 3 studies showed Nasal emission Event rate (%) (Proportion) 41.304% with significant heterogeneity p-value 0.0001. 6 studies showed speech therapy Event rate (%) (Proportion) 30.348% with significant heterogeneity p-value 0.0001. 3 studies showed hyper-nasality Event rate (%) (Proportion) 30.954% with insignificant heterogeneity p-value 0.2532

In addition, Shaffer et al.,¹² stated that all children received speech evaluations during the study period; 228 (98.3%) had evaluations after palatoplasty and before 36 months of age. At this evaluation, 124 (54.4%) of 228 had a speech/language delay and 11 (4.8%) of 228 had an ONF in the previously repaired area. Speech therapy was received or recommended for 135 (59.2%) of 228 children. Similarly, Ha et al.,¹⁷ showed that 50.3% of all the patients had received speech therapy and 28.8% and 51.4% demonstrated significant hypernasality and articulatory deficits, respectively.

The present study showed that 3 studies showed Perforation Event rate (%) (Proportion) 18.967% with significant heterogeneity p-value <0.0001. 3 studies showed Myringosclerosis Event rate (%) (Proportion) 25.134% with significant heterogeneity p-value 0.0456. 3 studies showed Retraction Event rate (%) (Proportion) 14.317% with significant heterogeneity p-value <0.0001. 2 studies showed Cholesteatoma Event rate (%) (Proportion) 3.497% with significant heterogeneity p-value 0.0014.

Imbery et al.,¹⁵ summarized the breakdown of complications by syndrome and cleft type. There was a total of 229 documented otologic complications, including myringosclerosis, retraction, perforations, ossicular abnormalities (eg, myringostapediopexy), and cholesteatoma in 197 unique patients. The current study showed that 2 studies showed poor hearing with tube Event rate (%) (Proportion) 43.327% with insignificant heterogeneity p-value 0.1278. Our results were supported by the study of Kim et al.,¹⁶ as they reported that after adjustment for potential confounders with multivariate analysis, patients who

received four or more sets of VTs, those with a history of TM perforation, and those with isolated cleft palate were found to be at an increased risk for hearing loss at 10-year follow-up. Age of first tube placement and total number of otologic complications were not independent predictors of hearing loss on adjusted analysis.

According to Yang et al.,¹³ a significantly higher percentage of poor hearing was also found in the children undergoing ventilation tube insertions two or more times ($p = 0.048$). Our results showed that 3 studies showed fistula Event rate (%) (Proportion) 8.482% with significant heterogeneity p -value 0.0230. 2 studies showed OM Event rate (%) (Proportion) 17.35% with significant heterogeneity p -value 0.0084. Our results were in line with study of Kobayashi et al.,⁵ as they reported that the clinical outcome after the age of 5 years (range 6-13 years, average 9.42 years) was investigated using the medical records of individual patients. The clinical course was rated as good in cases that remained free of OME even after the age of 5 years, or cases in which OME developed after the age of 5 years but healed in response to conservative treatment. The clinical course was rated as poor in cases who developed OME after the age of 5 years, necessitating further surgical treatments. The outcome at the final examination was rated as good for 56 ears (68.3%) which had been treated by VT placement, and for 95 ears (95%) that did not require VT placement.

In the study of Ha et al.,⁵ patients with CPO (clefts of palate only) appear to have a slightly higher rate of oronasal fistula development following primary palatal surgery (9.6%) compared to patients with UCLP (unilateral cleft lip and palate) or BCLP (bilateral cleft lip and palate) (7.3% and 5.3%, resp). However, Chi-square analyses revealed that the rate of oronasal fistula was not significantly different among the three groups of cleft type. The relationship between cleft type and percentage of patients who had received secondary palatal surgery was significant.

Conclusion

We have produced a recommendation about the outcomes that should be measured, as a minimum, in studies of the management of OME in children with CP. The development process included input from key stakeholders and used novel methodology to integrate the opinion of healthcare professionals, parents and children.

Conflict of interest: no conflicts of interest.

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